Psychosocial impact on families with an infant with a hypoplastic left heart syndrome during and after the interstage monitoring period - a prospective mixed-method study

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Abstract: AIMS AND OBJECTIVES To investigate parents' experiences, coping ability and quality of life while monitoring their sick child with hypoplastic left heart syndrome at home. BACKGROUND Interstage home monitoring for children with hypoplastic left heart syndrome reduces interstage mortality between Norwood stages I and II. Little is known about the psychosocial impact of interstage home monitoring. DESIGN Prospective mixed-method study. METHOD This study assessed the psychosocial impact on parents during IHM. This contains for quantitative assessment the Short Form Health Survey questionnaire and the Impact of Family Scale administered 1 and 5 weeks following discharge before and after stage II. For qualitative assessment semi-structured interviews focussing on the postdischarge coping strategies were conducted twice, 5 weeks after hospital discharge before and after stage II. RESULTS Ten infants (8 males) with hypoplastic left heart syndrome (n=7) or other types of univentricular heart malformations (n=3), and their parents (9 mother/father 2-parent households, 1 single mother) were included. There were no interstage deaths. Mental Health Composite Summary scores were low in both parents (mothers: 40.45±9.07; fathers: 40.58±9.69), and lowest for the item "vitality" (mothers: 37.0±19.46; fathers: 43.12±25.9) before and after stage II. Impact of Family Scale values showed higher daily and social burdens for mothers. "Becoming a family" was the most important task as coping strategy to equilibrate the fragile emotional balance. The parents judged interstage home monitoring as a protective intervention. CONCLUSIONS Although psychosocial burden before and after stage II remains high, becoming a family is an essential experience for parents and confirms their parenthood.

DOI: https://doi.org/10.1111/jocn.13694

Received Date : 20-Jul-2016
Revised Date : 22-Nov-2016
Accepted Date : 10-Dec-2016
Article type : Original Article

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The project was supported by the Anna Müller Grocholski Foundation (Reachout II)

This article has been accepted for publication and undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1111/jocn.13694

Conflicts of interest

The manuscript has been written by the first author. No honorarium, grant, or other form of payment was given to anyone to produce the manuscript. The authors declare no conflicts of interest.

Funding

The sponsor had no influence on study design, the collection, analysis and interpretation of data, the writing of the report and the decision to submit the paper for publication.

Acknowlegements

Many thanks to all participating parents, Rabia Liamlahi (Study nurse), Nicole Tracy Graf Johansen (Statistics) and Marcia Leventhal (Translations).

Short running title

Quality of life during interstage in hypoplastic left heart

Abstract

Aims and Objectives. To investigate parents' experiences, coping ability and quality of life while monitoring their sick child with hypoplastic left heart syndrome at home.

Background. Interstage home monitoring for children with hypoplastic left heart syndrome reduces interstage mortality between Norwood stages I and II. Little is known about the psychosocial impact of interstage home monitoring.

Design. Prospective mixed-method study.

Method. This study assessed the psychosocial impact on parents during IHM. This contains for quantitative assessment the Short Form Health Survey questionnaire

and the Impact of Family Scale administered 1 and 5 weeks following discharge before and after stage II. For qualitative assessment semi-structured interviews focussing on the postdischarge coping strategies were conducted twice, 5 weeks after hospital discharge before and after stage II.

Results. Ten infants (8 males) with hypoplastic left heart syndrome (*n*=7) or other types of univentricular heart malformations (*n*=3), and their parents (9 mother/father 2-parent households, 1 single mother) were included. There were no interstage deaths. Mental Health Composite Summary scores were low in both parents (mothers: 40.45±9.07; fathers: 40.58±9.69), and lowest for the item "vitality" (mothers: 37.0±19.46; fathers: 43.12±25.9) before and after stage II. Impact of Family Scale values showed higher daily and social burdens for mothers. "*Becoming a family*" was the most important task as coping strategy to equilibrate the fragile emotional balance. The parents judged interstage home monitoring as a protective intervention.

Conclusions. Although psychosocial burden before and after stage II remains high, becoming a family is an essential experience for parents and confirms their parenthood.

Relevance to clinical practice. Healthcare professionals must be aware of parents' needs during this vulnerable interstage period and to provide psychosocial and nursing support.

Key words: hypoplastic left heart syndrome, interstage monitoring, psychosocial impact, quality of life, family, infant

What does this article contribute to the wider global clinical community

- Healthcare professionals have an important role in supporting families of infants with hypoplastic left heart syndrome and can use the family assessment to evaluate the family resources.
- Comprehensive education will enable parents and family members to be involved in the care of their child and to become the child's experts.
- In addition to the practical care of their child, parents need to be provided with emotional and psychosocial support to proactively prepare them for the possible stressful situations and problems that might arise.
- Parents have to learn to use their own coping strategies and to be the advocate for the interests of their child when working with the multi-professional health care team.

Introduction

Hypoplastic left heart syndrome (HLHS) remains one of the most severe type of the congenital heart disease (CHD) with a frequency of 2:10,000 live births (Pradat *et al.* 2003). HLHS can be treated by cardiovascular surgery after birth with the stage I Norwood procedure followed by the stage II Norwood procedure later at age 3 to 6 months (Akintuerk *et al.* 2002, Barron *et al.* 2009). One of the most critical periods is the interstage period between stages I and II, associated with an interstage mortality up to 15% (Knirsch *et al.* 2014). But, interstage mortality can be reduced using interstage home monitoring (IHM) (Ghanayem *et al.* 2004, Ghanayem *et al.* 2003). Before discharge from hospital, *advance practice nurses* instruct parents for IHM to realise and manage observed clinical changes in all required care tasks. After discharge, IHM is then performed by the parents at home with daily measurement of

body weight and transcutaneous oxygen saturations. In case of clinical deterioration parents immediately report back to the heart centre to ensure early treatment to overcome interstage mortality. The impact on the quality of life of the parents and the burden of this additional responsibility due to IHM is not known.

Therefore, we determined quantitative and qualitative factors affecting the psychosocial impact of IHM by using a mixed-method study design. By that, we aimed to evaluate the psychosocial impact of IHM on the families, potential differences between father and mother on quality of life and their ability to cope during and after Norwood stage II.

Background

Cardiavascular surgery for HLHS aims to establish the systemic blood circulation with the right ventricle as the systemic pumping chamber by reconstructing the hypoplastic aortic arch combined with the pulmonary circulation with systemic-to-pulmonary shunt. For stage I the Norwood procedure or Hybrid procedure (by patent arterial duct stenting and balloon atrioseptostomy and bilateral pulmonary artery banding) is performed within the first week of life (Akintuerk *et al.* 2002). The stage II Norwood procedure or combined stage I/II procedure after Hybrid with bidirectional cavopulmonary anastomosis follows at age 3 to 6 months (Barron *et al.* 2009, Pradat *et al.* 2003).

Between stage I and II, this interstage period is crucially affected by a high interstage mortality. Up to 15% of the infants may die during this period because of hemodynamic compromise due to fluid volume shift caused by febrile respiratory or gastrointestinal infections. These hemodynamic changes may alter the coronary artery and pulmonary perfusion leading to myocardial ischemia, reduced cardiac

output or severe cyanosis in the case of shunt thrombosis (Ghanayem *et al.* 2004, McGuirk *et al.* 2006, Pradat *et al.* 2003). Interstage mortality is independent of the type of stage I procedure, i.e. Norwood or Hybrid procedure (Knirsch *et al.* 2014).

But, having a child born with HLHS presents a painful and stressful process for the involved families (Sarajuuri *et al.* 2012). Although IHM actively involves the parents in the at-home clinical management of their infants, it also means additional responsibility for the parents and potential load for the whole family living with a small child with HLHS.

Methods

Study design

This multiprofessional project used a prospective sequential mixed methods study design to analyse the psychosocial impact on parents performing IHM before and after the Norwood stage II procedure (Polit & Tatano Beck 2004). All parents of infants with HLHS undergoing the stage I (or hybrid) procedure between February 2011 and December 2012 were invited to participate in the study.

Inclusion criteria were families who were able to speak German, answer all of the questionnaires, and participate in the interview. Exclusion criteria were families of infants who could not be discharged home between stages I and II.

The study conforms to the principles outlined in the Declaration of Helsinki and was approved by the local ethical committee. All data including sociodemographic, health-related information and the interviews were anonymised for analysis.

A time-line of the quantitative and qualitative measurements is given in Table 1.

Quantitative assessment

Health-related quality of life

As a standardized measurement of perceived health we used the SF-36 questionnaire (Bullinger 2000). The SF-36 consists of eight scaled scores, which are the weighted sums of the questions in their section. The eight sections include physical functioning, role limitations due to physical health, bodily pain, general health perception, vitality, social functioning, role limitations due to emotional problems, and mental health. Each scale ranges from 0 (maximal disability) to 100 (no disability). For somatic health status the physical health component summary score (PCS) and for psychic health status the mental health component summary score (MCS) validated for the general German population (Ellert & Bellach 1999, Ellert & Kurth 2004), with Cronbach's $\alpha > 0.85$, Reliability coefficient > 0.75 for all areas except social functioning (α=0.73, reliability=0.74) (Brazier *et al.* 1992, Jenkinson *et al.* 1993).

Impact on Family Scale (IOF-G)

For analysis of the psychosocial impact on families living we used the IOF-G in its German version (Stein & Riessman 1980), validated in 2001 (Cronbach α = 0,88) (Ravens-Sieberer *et al.* 2001). The IOF-G consists of five psychosocial dimensions including problems in coping, social relationships, concern of siblings, general negative impact, and financial impact. Each dimension is scaled from 1 (lowest impact) to 4 (highest impact).

Qualitative assessment

Semi-structured interviews were conducted at two time points (T2 and T4). The first was during IHM, and the second was after the stage II procedure when IHM was no longer necessary (Table 1). The interviews were based on the postdischarge coping difficulty scale (Weiss *et al.* 2008). This 10-item scale includes difficulties with stress, recovery, self-care, and self-medical management; family difficulty; help and emotional support needed; confidence in self-care and medical management abilities; and adjustment. The scale was self-adapted to the specifics of families with HLHS patients and conducted by the first author at home. All interviews were digitally recorded, transcribed verbatim and anonymised.

Qualitative content analysis

All interviews were analysed using the technique of Mayring et al. (Mayring & Gläser-Zikuda 2008). This qualitative content analysis offers the opportunity to develop methods of systematic interpretation of the content of the interviews, which are applicable to the qualitative components necessarily involved in every content analysis, systematizing and making them testable through stages and rules of analysis. For the qualitative data analysis software Atlas.ti was used (Atlas.ti version 6.2, Scientific Software Development GmbH, Germany) to paraphrase, summarize and reduce the text in a sentence-by-sentence approach. The text was further condensed into generalized categories, summarizing the content as much as possible. Repetitive round table discussions with the coauthors were performed to ensure the reliability and validation of the analyses. Finally, the models were re-validated with three study families to ensure formal and content-related correctness.

Statistics

All data were analysed with the Statistical Package for Social Sciences version 20 (SPSS, IBM Corporation 2011, Chicago, USA). Descriptive statistics were used to determine the mean and standard deviation (SD) for the IOF-G and SF-36. As we had no normally distributed data we used non-parametric tests. The Friedman Test was used to compare time points T1 and T2, and T3 and T4. The Dunn's multiple comparison test was used for the comparisons of the two time points T1 / T3 and T2 / T4–For the comparison of mother and father, all mean values of the time points were-calculated and the Wilcoxon signed-rank test was performed.

Was A *p*-value <0.05 was judged as statistically significant.

Results

Demographic results

Nine 2-parent couples and one single mother were recruited for the interviews. Two couples declined participation due to lack of time. Mothers' median age was 29 (20-39) years and fathers' median age 32.5 (22-48) years. Seven mothers and six fathers were Swiss nationals. The infants were first born children in five of ten families. Three families had one child, one had two children, and one had three children.

The neonates were treated for HLHS between February 2011 and December 2012. Seven were diagnosed with HLHS and three had a variant of HLHS with univentricular physiology with shunt-dependent pulmonary perfusion. Seven couples were informed about the cardiac diagnosis during pregnancy. Eight infants were male. The mean duration of hospitalisation after stage I was 48.5±9.2 days, and after stage II was 31.6±13.9 days.

Health-related quality of life of the parents

The SF-36 with eight scaled scores which ranges from 0 (maximal disability) to 100 (no disability) showed in the comparison at all four points in time (T1-T4) that the PCS values for mothers (58.17±6.77) and fathers (60.02±3.92) were one standard deviation above the general German population (Ellert & Bellach 1999, Ellert & Kurth 2004) (Table 2). In contrast, the MCS values were lower for mothers (40.45±9.07) and fathers (40.58±9.69), but were comparable for both mothers and fathers at all four time points (T1-T4). The highest SF-36 values were found for physical functioning in fathers (99.39±1.16), while lowest values were found in vitality for mothers (43.23±17.13) and fathers (50.68±20.70). Two factors were associated with having an impact on MCS and PCS: prenatal diagnosis of CHD showed a trend towards higher MCS values (42.97±9.11 vs 34.10±5.41, p=0.059) and families with older siblings had lower PCS values (55.89±6.11 vs. 61.48±3.87, p=0.043).

Impact on Family

The IOF-G with the five psychosocial dimensions scaled from 1 (lowest impact) to 4 (highest impact) showed a substantial psychosocial burden with higher scores for mothers than fathers for the entire study period (T1-T4). This trend toward higher burden for mothers was universal in all dimensions except the financial impact, which was more pronounced in fathers. The highest scores were found in 'general negative impact' for both mothers and fathers (2.45±0.49 and 2.21±0.34, respectively, p<0.05) (Table 3), while mean scores below 2 were found for 'concern for siblings' (mother 1.93±0.31; father 1.68±0.27), 'problems with coping' (mother 1.69±0.36; father 1.48±0.16) and 'financial impact' (mother 1.64±0.58; father 1.90±0.49). Factors such as prenatal diagnosis of CHD, length of hospital stay,

families with older siblings or families from foreign nationality had no influence on the scores of the IOF-G.

Qualitative Results

"Becoming a family" after the first discharge

"Becoming a family" is the central goal for parents when they are first discharged to home after stage I. Parents look forward to this initial discharge to finally be a "real" family in their own home. A mother describes being at home as follows *"…we are enjoying being together, the first time we really feel like a family…"(P14-15).*

Emotional burden after the first discharge

"Emotional Burden" is described by parents as including a wide variety of emotions, such as the fear and insecurity experienced immediately following the first hospital discharge. Mothers found 'being alone with the child at home in the initial period' as stressful. *M: "Yes, what was really hard was that I was alone at home in the first week. That was really hard for me…Yes, you have the diagnosis and the insecurity.". (P3-99)* Parents evaluate IHM primarily as a security-enhancing measure. *M:"I think it would be even worse to go home without it (monitoring."(P15-251)*

The parents' chronic fatigue is one of most frequent symptoms attributed to their daily burden. Parents are awake at night either due to the fear of missing some change in the child's condition or because treatments are needed late at night or early in the morning. Parents also describe the ongoing stress that began when their child was first diagnosed with HLHS, as an additional cause of their chronic fatigue. Symptoms related to their chronic fatigue range from a lack of concentration to total

exhaustion. *M: " It's as if I (…) somehow forget what I am supposed to do with him, like almost being overwhelmed or (…) yes, very tired, exhausted."*(P14-13)

"Being a family" following discharge after stage II

Parents describe "being a family" and experiencing a "new normality in their everyday life", following discharge after the stage II procedure. It allows the family a new start after the intense and stressful first months.

Emotional burden after stage II

It was only after stage II ended that some parents realized how stressful the past months had been. They describe having had to expend a great amount of energy to adjust to their new normality. They feel relieved that their child is doing better and that they are not continually confronted with the illness.

After stage II, parents often experience their child as more active and are excited about the positive developmental steps that come with these changes. During this phase, the child's illness plays a secondary role and questions about nutrition, weaning from the feeding tube, upbringing and neurodevelopment come into the foreground. Parents begin to allow themselves to make more concrete plans for the future. *M: "And we finally have time for his development and (....) to think about how we want to raise him, that really wasn't an issue before. So (…) we didn't have time to think about that, and now we actually deal with these normal thing." (P11-36)*

Parents are constantly challenged by the unpredictable course of their child's illness and find themselves in a fragile balance between "becoming a family" and struggling with their "emotional burden".

Discussion

This prospective mixed methods study is the first to analyse the impact on quality of life, coping and burden for parents and families taking care of their newborn with HLHS in the first months of the child's life. Families with a child with HLHS are confronted with an intense, dynamic and stressful transformational process from diagnosis until the hospital discharge after

stage I.

Interestingly, the physical health of parents with a firstborn child with HLHS was better than in the general German population (Ellert & Bellach 1999, Ellert & Kurth 2004). In contrast, parents who had other children showed lower physical health (p=0.043). Regarding mental health, mothers and fathers scored lowest for "vitality" from T1 to T4, although fathers showed a slight increase in scores over time. These results correlated with the interviews where constant fatigue was described as the most burdensome symptom

Of note, the group of parents who had had a prenatal diagnosis tended to show a higher MCS score. This could be an indicator that in spite of the burden of the prenatal diagnosis, the time leading up to the birth is important for parents to prepare themselves for the child's complex illness after birth (McKechnie & Pridham 2012) and show an increased understanding of their child's disease at discharge (Williams *et al.* 2008).

Mothers have higher family burden, especially the daily and social burdens (Table 3), indicating that mothers feel primary responsibility in caring for the child and the family. In contrast, fathers showed higher financial burden (Table 3), probably related to the traditional father's role as the sole wage earner, and to increased pressure from missing work due to the child's illness. A decrease in family burden from before

to after stage II Norwood procedure was not found in our study. We found an increase in burden between T1 and T2 as shown in the item “coping difficulties”.

One previous study examined the impact of having an infant with severe CHD on family life one year after surgery. No significant differences between mothers and fathers in the total score using the IOF-G were found (Werner *et al.* 2014). The differences we found in our small cohort may be interpreted as an acute effect on family burden focused on the first months of life and can be interpreted due to the unpredictable course of the life-threatening illness and the demanding care tasks at home. A significant relationship was found between the age of the child, type and severity of CHD, associated comorbidities, financial situation and number of older siblings (Arafa *et al.* 2008). Higher stress, hopelessness and social isolation of the parents is associated with lower quality of life, and additionally, the mothers rated their quality of life lower than the fathers (Lawoko & Soares 2002).

Regardless of the uncertainty, parents desire to be at home with their child (Rempel & Harrison 2007).

The first discharge from hospital is essential for the family and results in the chance for them to "become a family" at home. Parents greatly appreciated this time at home in spite of the many challenges, thus the term "parenting under pressure", that is used for parents with HLHS children (Rempel *et al.* 2013). Parents need the best training and support to cope during this demanding time. This includes optimal and early training well before the child is discharged after the stage I procedure. A close multiprofessional collaboration between the advanced practice nurses and the pediatric cardiologists is advantageous in building a supportive network in the outpatient setting for the interstage period. Training in practical IHM skills includes measuring body weight and transcutaneous oxygen saturation, documentation,

interpreting and managing the observed clinical changes, and performing all the required care tasks.

Parents without family support and single mothers experienced a greater burden over a longer time and needed help and assistance earlier. An evaluation of the family resources and the parents' roles will indicate the need for additional care. The received social support can serve as a resilience factor between the daily stress and parental coping (Tak & McCubbin 2002). Knowledge of the magnitude of the family burden forms the basis to implement early psychosocial and nursing interventions aimed at preventing negative implications for the child and his family (Werner *et al.* 2014).

Parents described "being a family" as an intensive developmental family process in gaining increasing competence after the interstage-period (Rempel *et al.* 2012). Only then would parents allow themselves to make concrete future plans for their families.

Throughout the critical first months of life, healthcare professionals need to anticipate the potential long-term outcomes that could result in additional challenges for the parents. These outcomes may be affected by the persistent stress on the family, or the stress on the child from the multiple surgical procedures, potentially resulting in neurodevelopmental and behavioural impairments (Brosig *et al.* 2007). Healthcare professionals should be sensitive to the high emotional burden and the possible differences in parenting roles in the critical first months, and provide low-threshold interventions.

Limitations

We included only a small, heterogeneous group of parents in terms of family situation, nationality, language and culture, and date of diagnosis and disease

progression, limiting generalisability. In this study, the mothers were the main caregivers and thus the study may lack data on the experiences of the fathers. Due to the low prevalence of babies born with HLHS, it would be advantageous to test these results in a large, multicentre longitudinal study using a mixed methods design, starting from the prenatal diagnosis until after stage III.

Conclusions

The diagnosis of HLHS in a child is associated with far-reaching changes in the life and future plans of the parents. The psychosocial burden before and after stage II remains high, but becoming a family is an essential experience for parents and confirms their parenthood. Health professionals should be aware of the vulnerability of the parents during the interstage period and provide psychosocial and nursing support. Furthermore, it is recommended to reflect critically on the previous division between inpatient and outpatient care for these patients and their families, especially during the interstage period. New models of care must include continuity of care at home and support for parents during this very challenging and important phase of "becoming parents".

Relevance to clinical practice

Despite confronted with a severe cardiac diagnosis of HLHS, families can be introduced performing IHM offering an opportunity to reduce interstage mortality. But, all healthcare professionals including *advance practice nurses* must be aware of parents' needs during this vulnerable interstage period and provide complex psychosocial and nursing support. Comprehensive education will enable parents and family members to be involved in the care of their child and to become the child's

experts. In addition to the practical care of their child, parents need to be provided with emotional and psychosocial support to proactively prepare them for the possible stressful situations. Parents may develop their own coping strategies by becoming a family.

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Tables

Table 1. Overview of time points (T1 to T4) for quantitative and qualitative assessment of psychosocial impact on families of an infant with HLHS.

Discharge after stage I procedure *with* interstage home monitoring		**Discharge after stage II procedure *without* interstage home *monitoring***	
One week after discharge (T1)	**Five weeks after discharge (T2)**	**One week after discharge (T3)**	**Five weeks after discharge (T4))**
IOF-G **SF-36**	**IOF-G** **SF-36** **Interview**	**IOF-G** SF-36	**IOF-G** SF-36 **Interview**

IOF-G, Impact on Family Scale-German version,
SF-36, Short Form Health Survey questionnaire

Table 2. Comparison of scores of SF-36 from mothers and fathers at all four points in time (T1-T4) before and after stage II procedure for HLHS with or without interstage home monitoring

	Mother (n=10)	**Father (n=10)**	**p value**
Physical functioning (n=8)	97.86 ± 2.99	99.38 ± 1.16	0.438
Role limitations due to physical health (n=8)	86.20 ± 23.12	88.80 ± 20.38	0.563
Bodily pain (n=8)	88.13 ± 11.42	90.56 ± 11.97	0.219
General health perception (n=8)	79.44 ± 19.15	84.59 ± 10.30	0.469
Vitality (n=8)	43.23 ± 17.13	50.68 ± 20.70	0.313
Social functioning (n=8)	81.90 ± 14.55	75.00 ± 19.55	0.156
Role limitations due to emotional problems (n=8)	73.96 ± 33.76	76.39 ± 22.02	0.750
Mental health (n=8)	66.17 ± 15.56	71.33 ± 14.64	0.148
PSC (n=8)	**58.17 ± 6.77**	**60.02 ± 3.92**	**0.844**
MCS (n=8)	**40.45 ± 9.07**	**40.58 ± 9.69**	**0.195**

PCS / MCS = T-values with mean 50 ; SD 10

Table 3. Comparison of values of IOF-G in five dimensions for mothers and fathers at all four points in time (T1-T4) before and after stage II procedure for HLHS with resp. without interstage home monitoring

	Mother (n=10)	**Father (n=10)***	**p value**
Problems in coping (n=8)	1.69 ± 0.36	1.48 ± 0.16	0.094
Social relationships (n=8)	2.45 ± 0.53	2.15 ± 0.62	0.063
Concern for siblings (n=3*)	1.93 ± 0.31	1.68 ±0.27	0.250
General negative impact n=8)	2.45 ± 0.49	2.21 ±0.34)	**0.047**
Financial impact (n=8)	1.64 ± 0.58	1.90 ± 0.49	0.086
Total score	**2.17 ± 0.39**	**2.00 ± 0.26**	**0.055**

Data are given as mean ± standard deviation.

Scale IOF-G: 1 = lowest, 4 = highest impact

* = calculated only for siblings at school age